Endobronchial metastasis from oral fibrosarcoma 13 years after treatment of primary tumor

Mohammad Hossein RAHIMI-RAD[1], Behrouz ILKHANIZADEH[2]

[1] Urmia Üniversitesi Tıp Fakültesi, İç Hastalıkları Bölümü, Urmia, Batı Azerbaycan, İran,
[2] Urmia Üniversitesi Tıp Fakültesi, Patoloji Bölümü, Urmia, Batı Azerbaycan, İran.

ÖZET

Primer tümör tedavisinden 13 yıl sonra gelişen oral fibrosarkomun endobronşiyal metastazı

Endobronşiyal metastazlar nadir görülür; böbrek, meme ve kolorektal kanserlerde daha fazladır. Diğer bildirilen primer tümörler melanom, sarkomlar, uterus, serviks, over, prostat, tiroid, pankreas ve adrenal bezlerdir. Literatürü gözden geçirdiğimizde sadece bir tane fibrosarkomun endobronşiyal metastazının (ispanyolca) bildirildiğini gördük. Primer tümör tedavisinden 13 yıl sonra lokal rekürrens gelişen oral fibrosarkomun endobronşiyal metastazı saptanan 56 yaşında kadın hastayı bildiriyoruz. Eğer geçmişte malignite öyküsü olan bir hastada bronşiyal tümörle uyumlu semptomlar varsa, arada 13 yıllık bir zaman olsa da, santral hava yolu metastazı olasılığının akılda tutulması gerektiğini belirtmek istiyoruz. Endobronşiyal metastazların birçok varsayılan mekanizmalarından birisi de oral kanserlerden direkt aspirasyon ve tümör hücrelerinin implantasyonudur.

Anahtar Kelimeler: *Endobronşiyal metastaz, atelektazi, pulmoner metastaz.*

SUMMARY

Endobronchial metastasis from oral fibrosarcoma 13 years after treatment of primary tumor

Mohammad Hossein RAHIMI-RAD[1], Behrouz ILKHANIZADEH[2]

[1]Department of Internal Medicine, Faculty of Medicine, Urmia University, Urmia, West Azerbaijan, Iran,
[2] Department of Pathology, Faculty of Medicine, Urmia University, Urmia, West Azerbaijan, Iran.

Yazışma Adresi (Address for Correspondence):

Dr. Mohammad Hossein RAHIMI-RAD, Bronchoscopy Unite, Imam Khomeini Hospital, Urmia, West Azerbaijan, Urmia- IRAN
e-mail: rahimirad@umsu.ac.ir

Endobronchial metastasis (EBM) is uncommon and frequently is seen in renal, breast, and colorectal carcinomas. Other reported primary tumors include melanoma, sarcomas, and tumors of the uterine cervix, testis, ovary, prostate, thyroid, pancreas, and adrenal glands. With reviewing the literature, we were able to find only one report of EBM from fibrosarcoma (in Spanish). We described a 56-year-old woman with EBM of oral fibrosarcoma with local recurrence 13 years after treatment of primary tumor. We conclude that the possibility of central airway metastasis should be kept in mind if patients with a past history of malignancy present with symptoms consistent with bronchial tumors, even if there are 13 years interval. Of several mechanisms EBM, we assume direct aspiration and implantation of tumor cells to bronchus from oral cancer.

Key Words: *Fibrosarcoma, endobronchial metastasis, atelectasis, pulmonary metastasis.*

Endobronchial metastasis (EBM) is defined as documented non-pulmonary neoplasms metastatic to the subsegmental or more proximal central bronchus, in a bronchoscopically visible range (1). It is the rarest form of intrathoracic metastasis from extrathoracic malignancies (2). The incidence of EBM estimated to be around 2% (3). However, its incidence may be underestimated because routine bronchoscopic examination is not commonly performed in patients with pulmonary metastasis (1).

Tumors more likely to give EBM are renal, breast, and colorectal carcinomas. Other reported malignancies include hepatocellular, ovarian, thyroid, uterine, testicular, nasopharynx, pancreas, prostat and adrenal carcinomas, sarcomas, melanomas, plasmacytomas, Wilms tumor and urinary bladder cancer (2-10). Fibrosarcoma is a malignant neoplasm of mesenchymal origin. It is a very rare malignancy but may occur anywhere in the body, in any age group and even as a congenital neoplasm. It arises from superficial and deep connective tissue (11).

We searched medline for keywords "Endobronchial metastasis" and fibrosarcoma on 19 March 2009. The result was only one report of EBM from fibrosarcoma in Spanish language in a patient with neurofibromatosis by Urrutia A et al. (12). With reviewing literatures, we were unable to find any other reported case of EBM from fibrosarcoma.

CASE REPORT

A 56-year-old woman presented to our clinic with dyspnea, cough and hemoptysis on 27 January 2008. She had past medical history of treatment for fibrosarcoma in left side of mouth in 1966. Her chest X-ray taken in another clinic on 10 January 2008 showed total left lung collapse (Figure 1A). Computerized tomography of chest (on 25 December 2007) revealed collapse of left lower lobe (Figure 1B). Physical examination revealed a woman without distress with relatively well condition. There was a red mass in left side mouth (Figure 2A). There was no lymphadenopathy. Trachea was deviated to left with decreased breath sounds on left hemithorax. Fibroptic bronchoscopy showed round mass in left main bronchus on Fi-

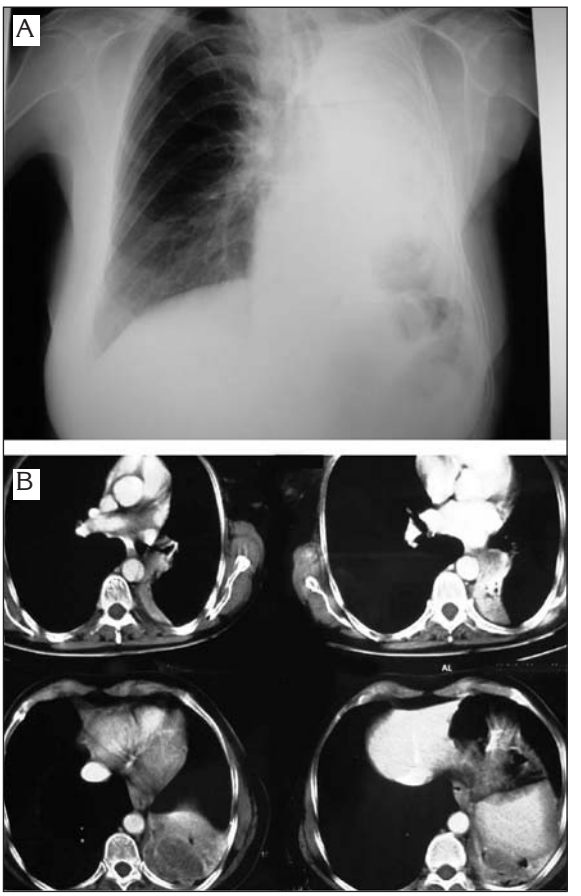

Figure 1. (A) Chest X-ray on shows left lung collapse, (B) earlier computed tomography scan on reveals left lower lobe collapse.

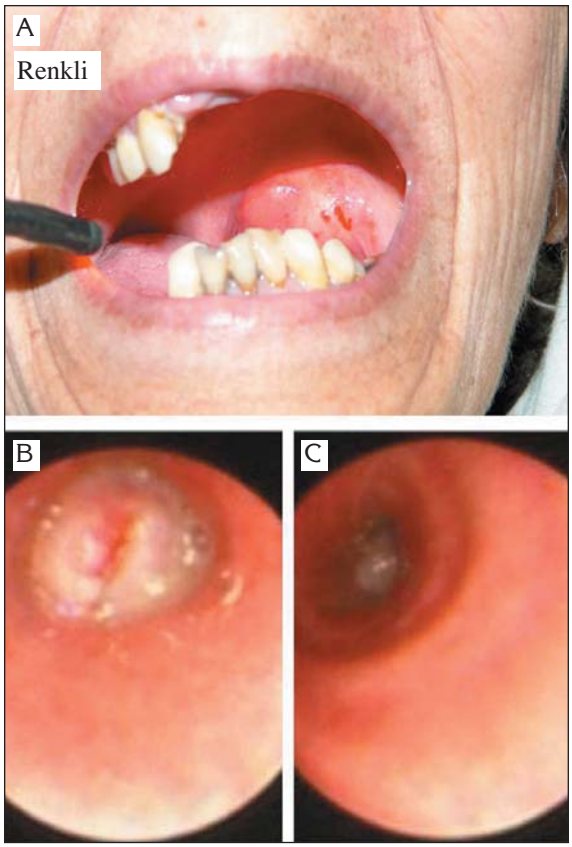

Figure 2. (A) Tumor in left side of mouth illuminated with bronchoscope; tumor in left main bronchus near view (B) and far view (C).

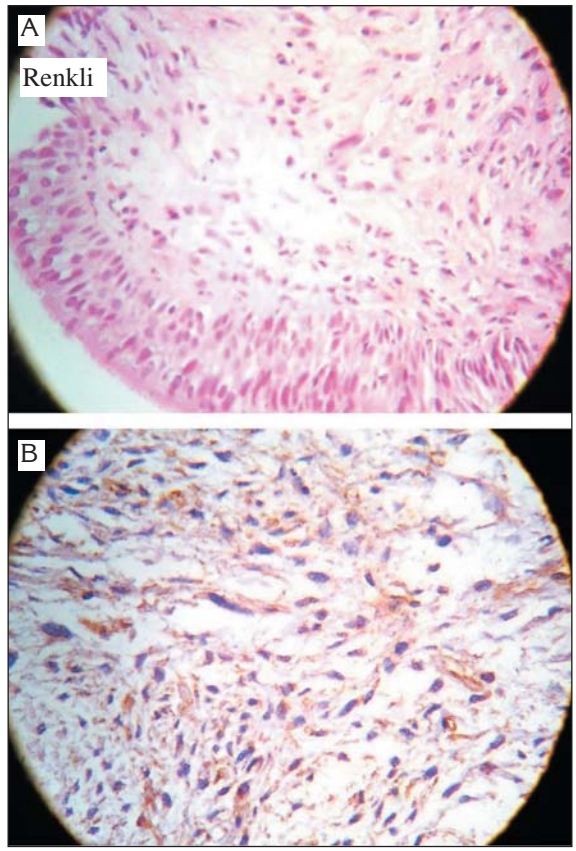

Figure 3. Histopathology of biopsy of left main bronchus mass shows (A) intact respiratory epithelium with intramural tumor consist of fibroblasts (HE x400) and (B), immunohistochemical staining with Vimentine is positive.

gure 2B and 2C and biopsy of mass confirmed fibrosarcoma (Figure 3). Immunohistochemical staining was positive for vimentin, and negative for S-100 protein, epithelial membrane antigen, smoth muscle actin, and chromogranin.

DISCUSSION

We presented a case of oral fibrosarcoma with EBM 13 years after diagnosis and treatment of the primary tumor with local recurrence. The time interval from the diagnosis of primary cancer to EBM diagnosis varies greatly. Sørensen reported that the mean time from diagnosis of initial cancer to the diagnosis of EBM was 50 months (range, 0-300 months)(13). However in some reports EBM were diagnosed before primary cancer diagnosis (14). It may be bilateral. Kanzaki et al. reported a case of bilateral EBM in post-operative synchronous adenocarcinoma of lung and stomach (15).

Four routes for EBM are suggested by Kiryu et al. (1):

1. Direct metastasis to the bronchus,

2. Bronchial invasion by a parenchymal lesion,

3. Bronchial invasion by mediastinal or hilar lymph node metastasis,

4. A peripheral lesion extended along the proximal bronchus.

The other suggested rout is aspiration of tumor cells from pharyngeal, tracheal, or other bronchial lesions (16). In our case, it was difficult to determine which mechanism pre-dominated; however, we assume that it was caused by direct aspiration and implantation of tumor cells to bronchus from mouth.

Eipe and colleagues suspected for direct tumor implantation into lower airways during anesthe-

sia in a patient with EBM following tongue cancer surgery (17). Were tumor cells implanted during previous surgery of tumor in mouth? Maybe, but there were 13 years interval, and it is not easy to conclude that hypothesis.

In our case the metastatic tumor was localized to the left main bronchus and lead to total collapse of left lung which was indistinguishable from primary bronchogenic carcinoma differential diagnosis of EBM is important and is influenced by an accurate clinical history and bronchoscopic investigation. These tumors may give rise to symptoms identical to primary bronchial carcinoma or carcinoid (8,18). The most frequent symptoms of EBM are cough, dyspnea and hemoptysis, however 52% of patients were asymptomatic in a study by lee et al. (19).

The roentgenographic findings in EBM are variable, atelectasis is the most common, followed by signs of multiple pulmonary nodules, perihilar masses, mediastinal lymphadenopathy or normal chest X-ray (5). It is in differential diagnosis with primary bronchogenic carcinoma and benign endobronchial vegetations (20,21).

Isolated EBM of fibrosarcoma is rare with only one case reports to date, we report the second one. The possibility of central airway metastasis should be kept in mind if patients with a past history of malignancy present with symptoms consistent with bronchial tumors, even if there are 13 years interval. Of several mechanisms EBM, we assume direct aspiration and implantation of tumor cells to bronchus.

REFERENCES

1. *Kiryu T, Hoshi H, Matsui E, et al. Endotracheal/endobronchial metastases: Clinicopathologic study with special reference to developmental modes. Chest 2001; 119: 768-75.*
2. *Lee KY, Ryu SJ, Joo M. Endobronchial metastasis of hepatocellular carcinoma. Yonsei Med J 2003; 44: 544-7.*
3. *Nair S, Kumar P, Ladas G. Intratracheal metastasis secondary to soft tissue liposarcoma. Singapore Med J 2007; 48: 81-3.*
4. *Erkal HS, Özturk AS, Kutluay L, et al. Endobronchial metastases from a leiomyosarcoma of the uterus. Tuberk Toraks 2008; 53: 74-8.*
5. *Gerogianni I, Gravas S, Papadopoulos D, et al. Endobronchial metastasis from prostate cancer. Int Urol Nephrol 2008; 40: 961-4.*
6. *Ulger Z, Karaman N, Piskinpasa SV, et al. Endobronchial metastasis of thyroid follicular carcinoma. J Natl Med Assoc 2006; 98: 803-6.*
7. *Shepherd MP. Endobronchial metastatic disease. Thorax 1982; 37: 362-5.*
8. *Mogulkoc N, Goker E, Atasever A, et al. Endobronchial metastasis from osteosarcoma of bone: Treatment with intraluminal radiotherapy. Chest 1999; 116: 1811-4.*
9. *Lee TP, Tzao C, Liu JH, et al. Isolated endobronchial metastasis of Wilms' tumor. J Pediatr Surg 2005; 40: 33-5.*
10. *Sakar A, Gencer N, Demireli P, et al. Endobronchial metastasis from urinary bladder cancer. Tuberk Toraks 2005; 53: 75-9.*
11. *Rosai J. Rosai and Ackerman's Surgical Pathology. 9th ed. Edinburg: Mosby, 2005.*
12. *Urrutia A, Guarga A, Tor J, et al. Endobronchial metastasis of a fibrosarcoma in multiple neurofibromatosis. Med Clin (Barc) 1983; 80: 600.*
13. *Sorensen JB. Endobronchial metastases from extrapulmonary solid tumors. Acta Oncol 2004; 43: 73-9.*
14. *Kim YS, Chang J, Shin DH, et al. Endobronchial metastasis of uterine cervix cancer: A two case reports and a review of the literature. Yonsei Med J 2002; 43: 547-52.*
15. *Kanzaki M, Onuki T, Tatebayashi T, et al. Bilateral endobronchial metastasis in postoperative stage I pulmonary adenocarcinoma. Diagn Ther Endosc 2000; 6: 141-5.*
16. *Yokoba M, Nishiia Y, Hagiria S, et al. Endobronchial metastasis from slow-growing lung cancer: A rare case report and review of the literature. Respir Med CME 2008; 1: 107-10.*
17. *Eipe N, Dildeep A, Samuel T. Endobronchial metastasis: An anesthetic complication? Can J Anaesth 2005; 52: 550-1.*
18. *Heitmiller RF, Marasco WJ, Hruban RH, et al. Endobronchial metastasis. J Thorac Cardiovasc Surg 1993; 106: 537-42.*
19. *Hanta I, Akcali S, Kuleci S, et al. A rare case of hurthle cell carcinoma with endobronchial metastasis. Endocr J 2004; 51: 155-7.*
20. *Rad MH, Alizadeh E, Ilkhanizadeh B. Recurrent laryngeal papillomatosis with bronchopulmonary spread in a 70-year-old man. Tuberk Toraks 2007; 55.*
21. *Rad MH, Milani M. Primary endobronchial actinomycosis simulating endobronchial tuberculosis in a patient with diabetes mellitus. Tuberk Toraks 2007; 55: 186-90.*